Polyarthritis and bone lesions complicating traumatic pancreatitis in two children

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The association of bone lesions, polyarthritis and cutaneous nodules with pancreatic disease is being recognized and reported more frequently. In adults all forms of pancreatitis and carcinoma of the pancreas have been involved, but in the few children described these complications have been associated with acute traumatic pancreatitis.

This paper describes two cases of acute traumatic pancreatitis in which polyarthritis and limb pains were noted after 2 to 3 weeks. In one child osteolytic lesions and periostitis were seen on roentgenograms 7 weeks after the onset of pancreatitis. In the other child minor roentgenographic changes were not seen until 5 months after the onset; however, bone scans showed clear-cut abnormalities after 1 month. Almost complete resolution could be expected within a year. Serum lipase and amylase concentrations remained elevated during the acute illness. Disseminated fat necrosis is apparently related to the excess amounts of circulating lipase.

L'association de lésions osseuses, de polyarthrite et de nodules cutanés avec une maladie du pancréas est reconnue et signalée plus fréquemment. Chez les adultes toutes les formes de pancréatite et de carcinome du pancréas ont été impliquées, mais chez les quelques enfants décrits ces complications ont été associées à une pancréatite traumatique aiguë.

Cet article décrit deux cas de pancréatite traumatique aiguë où une polyarthrite et des douleurs des membres sont apparues après 2 à 3 semaines. Chez un enfant des lésions ostéolytiques et une périostite ont été vues à la roentgénographie 7 semaines après le début de la pancréatite. Chez l'autre enfant des changements roentgénographiques mineurs n'ont pu être observés que 5 mois après le début; toutefois, des scintigraphies osseuses montraient des anomalies évidentes après 1 mois. Une guérison presque complète peut être anticipée en moins d'un an. Les teneurs sériques en lipase est en amylase sont demeurées élevées durant la phase aiguë de la maladie. La nécrose adipose disséminée est apparemment reliée à un excès de lipase circulante.

The fact that bone lesions, polyarthritis and cutaneous nodules may be complications of pancreatic disease is not known by most physicians. There have been a number of reports of the occurrence of one or all of these complications,18 but only five affected children have been described11,12 and only two of these in an English-language pediatric journal.11,12 Two of the affected children had pancreatic disease associated with the "battered-child syndrome".11,12

We present two more cases of children with acute traumatic pancreatitis and such complications. In one of the children polyarthritis and osseous lesions developed. In the other polyarthritis and one tender subcutaneous nodule appeared; roentgenograms did not demonstrate bone changes for 5 months, but a bone scan showed evidence of widespread osseous involvement after 1 month.

Case reports

Case 1

A 6-year-old boy was admitted to hospital May 6, 1975. Two days prior to admission, after being kicked accidentally in the abdomen, he experienced abdominal pain immediately and vomiting 1½ hours later. The pain persisted until admission. As a newborn infant he had had an operation for duodenal atresia.

Slight abdominal distention and generalized tenderness were noted. A tentative diagnosis of traumatic pancreatitis was confirmed by the finding of an increased serum amylase concentration of 1400 U/dL. A nasogastric tube was inserted and therapy with intravenous fluids started. A laparotomy was indicated, but because the boy’s parents were Jehovah’s Witnesses and would not allow a blood transfusion he was treated conservatively.

The child did well until May 16, when intermittent abdominal pain and a fever developed. A pancreatic cyst was suspected but it was decided to defer operation for about 3 weeks to allow the cyst wall to thicken.

On May 28 the child complained of pain in his elbows and knees; although the elbows were swollen and tender no heat or redness was detected. By June 4 both elbows were swollen and the child complained of severe pain in all his joints and refused to walk. Roentgenograms made the next day showed a marked effusion in the right elbow and mild effusion in the left. The knees and ankles were normal. A painful left hydrocele was also noted. Throughout this period his temperature remained elevated. By June 11 the hemoglobin concentration had decreased to 8.2 g/dL and the erythrocyte sedimentation rate was 121 mm/h (Westergren). The serum amylase value was 8000 U/dL.

At operation the next day the abdominal cavity was found to contain about 500 mL of cloudy fluid similar to that aspirated from the left hydrocele. A pancreatic cyst about 10 cm in diameter was leaking in the centre; it was marsupialized to the upper part of the wound because it was too thin to be Anastomosed to the bowel. The amylase concentration of the abdominal fluid was 70,000 U/dL. The child’s general condition improved greatly after the operation but the arthritic changes persisted, eventually subsiding.

Roentgenograms made June 22 showed that the fluid in the elbows had partly disappeared and there was evidence of subperiosteal new bone formation around the distal humeri and the upper ends of the ulnae and radius. About this time the child had begun to complain of pain in his shins; there was pronounced tenderness along both tibiae. The same day roentgenograms showed bilateral subperiosteal new bone formation affecting both tibiae and fibulae and mottled radiolucent lytic changes within the diaphysis of the right and left tibiae similar to those of multifocal osteomyelitis (Fig. 1).

On July 24 the dorsum of the right hand had become red and puffy, especially over the third metacarpal. Roentgenograms showed subperiosteal new bone formation along the shafts of the right second, third and fifth metacarpals and early bone lysis of the right second and third metacarpals (Fig. 2).

On July 15 the child still walked carefully and stiff-legged, but his condition continued to improve slowly and by July 30 his elbows and hands...
were normal though the left shin was still tender. On Aug. 5 anteroposterior views of the bones of the arms and legs showed extensive subperiosteal new bone formation involving the humeri, radii, right metacarpals, ulnae, tibiae and fibulae (Figs. 3 and 4). By Aug. 29 the child had no pain in the joints or bones. Roentgenograms made on Nov. 7 showed striking improvement, but there was still subperiosteal new bone formation, particularly along the shafts of the tibiae. By June 28, 1976 there was only minimal cortical thickening where previously there had been subperiosteal new bone formation of the distal medial shaft of the humeri.

**Case 2**

A 13-year-old boy was admitted to hospital Sept. 2, 1976. He had been injured by a knee blow to his upper abdomen and was complaining of pain in the left side of his abdomen and in his left shoulder. The splenic area and left costovertebral angle were very tender. The leukocyte count was 10.0 \( \times 10^9/\text{L} \), with 92% neutrophils, and the serum amylase concentration was 1265 U/dL.

At operation the same day a retroperitoneal hematoma was found on the left side and swelling and contusion over the pancreas with signs of traumatic pancreatitis. Drainage tubes were inserted before the incision was closed. Postoperatively the boy had a fluctuating fever and continuing abdominal tenderness.

About Sept. 14 he began complaining of pain in his limbs and joints, particularly in his left foot along the fifth metatarsal, where puffiness and redness were noted. Three days later he complained of pain in his right foot and there were similar findings. He also had soreness over the calcanea and ankles with tenderness and puffiness, but his ankle movements were normal.

On Sept. 18 a tender, red, indurated area about 1 cm in diameter was noted in the lower part of his right calf. This regressed after a week and was almost gone by 2 weeks. No other such lesions appeared. His temperature continued to spike to 39°C until Sept. 26, when it became normal.

On Sept. 28 the boy complained of soreness over his left hip and greater trochanter; the area was tender but not swollen. He continued to have some tenderness over the left fifth metatarsal. His weight had decreased to 33 kg from 40 kg preoperatively. The pain in his hands and feet decreased after a few days, his general condition gradually improved and his abdominal pain subsided.

On Oct. 1, 1976 bone scanning was...
done after administration of 10 mCi of technetium-99m methylene diphosphonate. The most easily recognized abnormality was an area of increased activity in the midshaft of the left tibia (Fig. 5). The other areas of increased activity were close to areas of normal epiphyseal activity and therefore the increased activity was not so pronounced. There was increased activity in the greater tuberosity of the left femur and probably in the upper metaphysis of the left radius, as well as in the right third, fourth and fifth and the left third and fourth metacarpals (Fig. 5). In comparison there was virtually no activity in the right first and

FIG. 4—Case 1, Aug. 5, 1975: more extensive subperiosteal new bone formation in right metacarpals.

FIG. 5—Case 2, Oct. 1, 1976: increased activity in right third, fourth and fifth and left third and fourth metacarpals and in left tibia.
second and the left first and fifth metacarpals, including the epiphyses.

The areas of increased activity presumably represented sites of bone infarction that were in the process of being repaired. Since there was virtually no activity in four of the metacarpals these bones may have been avascular at this stage.

On Oct. 2 the boy complained of pain in the first interphalangeal joint of his right index finger, which was puffy and tender but not swollen. On Oct. 7 and 13, after he complained of pain in the joints, roentgenograms of the chest, tibiae, fibulae, feet and hands showed no evidence of bone changes.

He was discharged Oct. 17 and was followed up as an outpatient. On Oct. 26 roentgenograms of the left femur, tibiae, fibulae, hands and feet were still considered normal. In mid-November the lateral aspect of his right hand was puffy and tender for 2 days.

Bone scanning was repeated Dec. 7, 1976. The midshaft of the left tibia now showed minimal activity (Fig. 6), which indicated that repair was almost complete. However, there was again intense activity in the greater tuberosity of the left femur, extending into the neck, which indicated that repair was continuing. The changes in the left radius had regressed. Increased activity was apparent in the shaft of the right third metacarpal. The right first and second and the left fifth metacarpals again showed reduced activity, although the activity in the epiphyses was now normal (Fig. 6). All other sites in the hands showed normal activity, which suggested that repair was complete. There was an ill-defined area of reduced activity in the region of either the anterior talus or navicular bone that had not been present previously; this probably represented another avascular site.

Roentgenograms of the hands and feet made Feb. 14, 1977 showed periosteal new bone formation around the right third and fifth metacarpal shafts.

Discussion

We believe that most physicians are unaware of the bone lesions, polyarthritis and panniculitis that may be associated with pancreatitis. When the arthritic lesions first appeared in patient 1 we considered that the child might have a collagen disease such as rheumatoid arthritis; the association of these lesions with traumatic pancreatitis was not appreciated then. Soon afterwards we became aware of the 1965 report by Schrier, Melmon and Fenster of subcutaneous nodular fat necrosis in an adult with pancreatitis. Further review of the literature disclosed a substantial number of reports, each describing one or two patients with pancreatic disease and one or more of these complications. Most textbooks and many reviews of pancreatitis, especially traumatic pancreatitis, did not mention these complications even though bone marrow involvement in a fatal case of pancreatitis was reported by Ponfick in 1872.

In their review of the literature in 1968 Mullin and colleagues described 23 patients with pancreatic disease and arthritis or skin lesions resembling erythema nodosum, or both, some of whom had bone lesions as well. Fourteen cases of carcinoma of the pancreas were associated with subcutaneous fat necrosis or joint involvement (12 with arthritis) or both. Of the 23 patients 2 had traumatic pancreatitis. The only child, aged 11 years, was the same one described by Immelman and associates in 1964; roentgenograms showed multiple osteolytic lesions in the radii and ulnae.

We were able to find only five other cases in the literature of pancreatitis and osteolytic lesions in children. In all instances the pancreatitis was traumatic. The second child was also referred to in a letter by Keeney, who enlarged on the child abuse spectrum. In this child more than 300 lytic lesions of the bones of the feet and hands as well as of several long bones were detected over a 3-week period. The lesions had almost completely healed 14 months later.

In one issue of the Journal of Rheumatology in 1975 Moore discussed syndromes resulting from dissemination of pancreatic enzymes, while Gibson and Tannenbaum and their colleagues discussed the association of bone lesions, polyarthritis and skin lesions with pancreatic disease. The consensus was that the disseminated fat necrosis that may affect viscera and bones distant from the pancreas is a result of the release into the bloodstream of excessive amounts of lipase because of obstruction of pancreatic outflow by tumours, calculi or edema or because of hypersecretion of functional acinar enzymes. The only case of traumatic pancreatitis in the literature was that of a child with bone lesions. The patient had a history of child abuse and was referred to Keeney, who thought that the bone lesions were due to compression of the pancreas by the head of the femur. In the case we report the midshaft of the left tibia showed minimal activity, which indicated that repair was almost complete. However, there was again intense activity in the greater tuberosity of the left femur, extending into the neck, which indicated that repair was continuing. The changes in the left radius had regressed. Increased activity was apparent in the shaft of the right third metacarpal. The right first and second and the left fifth metacarpals again showed reduced activity, although the activity in the epiphyses was now normal (Fig. 6). All other sites in the hands showed normal activity, which suggested that repair was complete. There was an ill-defined area of reduced activity in the region of either the anterior talus or navicular bone that had not been present previously; this probably represented another avascular site.

Roentgenograms of the hands and feet made Feb. 14, 1977 showed periosteal new bone formation around the right third and fifth metacarpal shafts.

FIG. 6—Case 2, Dec. 7, 1976: reduced activity in right first and second and left fifth metacarpals and minimal activity in midshaft of left tibia.
cell carcinomas and their metastases. Elevated serum concentrations of lipase and amylase have been found whenever sought in these cases (Table I).

The pathologic findings in bone, periartritic tissue and skin have been well described in several articles. The only patient other than the second of ours to have bone scanning done was described by Gibson and associates. A 55-year-old man had increased activity in the proximal end of the right humerus, where an area of lucency was noted roentgenographically; this region had not been the source of symptoms. In our patient bone scans showed widespread changes 1 month after the onset of traumatic pancreatitis, whereas roentgenologic abnormalities were not detectable for another 4 months, and then were minimal and confined to two metacarpals of one hand. The sites of bone scan abnormalities do not necessarily coincide with the sites of symptoms and signs.

We believe it is important for physicians to be alert to the association of bone lesions, polyarthritis and subcutaneous nodules simulating those of Weber-Christian disease in cases of pancreatitis, whatever the cause. If one is not aware of these complications fruitless investigation and unnecessary treatment for osteomyelitis and rheumatoid arthritis may be undertaken. In eight other children seen previously with acute traumatic pancreatitis we had not noted these complications, but after dealing with patient 1 we had no difficulty in recognizing them in patient 2.

There is apparently no specific therapy for the condition. It appears to be self-limited, resolution of the pancreatitis being followed by complete healing of the bone and joint lesions. The bone changes have regressed almost completely within a few months to a year. Bone scans show lesions early despite the lack of roentgenologic evidence. No doubt, if followed up long enough, these changes also regress completely.

We thank Dr. A.A. Wilkinson, chief of nuclear medicine at University Hosptal, Saskatoon, for his cooperation and interpretation of the bone scans.

References


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Table I—Summary of pertinent laboratory data in two cases of pancreatitis complicated by bone lesions, polyarthritis and cutaneous nodules

<table>
<thead>
<tr>
<th>Date</th>
<th>Case 1 Onset of pancreatitis May 6, 1975</th>
<th>Case 1 Onset of pancreatitis Sept. 2, 1975</th>
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<tr>
<td>Serum amylase, U/dL (modified)</td>
<td>2100</td>
<td>534</td>
</tr>
<tr>
<td>Van Loon method; normal range of values 50 to 170</td>
<td>196</td>
<td>48</td>
</tr>
<tr>
<td>Serum lipase, Case 1, U/mL</td>
<td>May 6</td>
<td>Sept. 16</td>
</tr>
<tr>
<td>(Cherry method)</td>
<td>1400</td>
<td>( \geq 1000 )</td>
</tr>
<tr>
<td>(Crandall method)</td>
<td>196</td>
<td>48</td>
</tr>
<tr>
<td>(Aug. 28)</td>
<td>48</td>
<td>103</td>
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<tr>
<td>Erythrocyte sedimentation rate, mm/hr (Westergren)</td>
<td>18</td>
<td>3</td>
</tr>
<tr>
<td>method; normal range of values 0 to 15</td>
<td>Sept. 2</td>
<td>3</td>
</tr>
<tr>
<td>Serum total protein and albumin, g/dL (normal ranges of values 6.0 to 8.0 and 3.5 to 5.5)</td>
<td>20</td>
<td>5.6, 2.5</td>
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<td>(Aug. 29)</td>
<td>29</td>
<td>1.2, 2.8</td>
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*From Princeton Bromedix Inc; Princeton, New Jersey.